Clinical Case Reports

ISSN: 2675-3626

Left anterior and Middle cerebral artery stroke associated with Pituitary apoplexy in an adolescent male with Giant prolactinoma: A Case Report.

Omane Acheamfour Okrah, Emmanuel Kwadwo Adjei Osei[1], Michael Yankey, Joseph Kebo Awindago, Emmanuel Jackson, Patrick Bankah

Department of Neurosurgery, Korle-Bu Teaching Hospital, Accra, Ghana

Emmanuel Kwadwo Adjei Osei, MD

e-mail: adjei.emma170@gmail.com

Available at:
http://www.archpedneurosurg.com.br/

Background: Pituitary apoplexy is a life-threatening condition in which there is a hemorrhage or infarction of the pituitary gland. It can be characterized by the sudden onset of headaches, vomiting, visual impairment, endocrine deficits, and changes in consciousness. There are very few cases of pituitary apoplexy presenting as strokes in children. We report a rare occurrence of an ischemic stroke in the left anterior and middle cerebral artery territories, caused by pituitary apoplexy from a prolactinoma in a 13-year-old male which was managed medically.

Case Presentation: He presented with worsening headaches and visual acuity and later developed left oculomotor nerve palsy, right hemiplegia, and dysarthria. MRI and CT imaging confirmed an apoplectic pituitary macroadenoma with internal carotid artery encasement and infarction in the territory of the left middle and anterior cerebral arteries. The patient was treated using dopamine agonists, which led to a decrease in tumor size and serum prolactin levels without surgery.

Conclusion: This report emphasizes the need to promptly recognize pituitary apoplexy as a potential cause of stroke in children. It also illustrates successful medical management in these cases, which could be especially important in areas with limited resources

Keywords: Pituitary prolactinoma, Apoplexy, Cerebral infarction, Middle cerebral artery, Anterior cerebral artery

INTRODUCTION

Pediatric prolactinoma is rare, with an incidence of 0.1 per 1,000,000 population, and it accounts for <2% of all intracranial tumors. The first case was reported by Bailey in 1898.[1] Brougham et al. introduced the term “pituitary apoplexy” and described the clinical and pathologic findings in five patients whose post-mortems revealed hemorrhagic necrosis.[2]

Only five juvenile cases of pituitary tumor apoplexy-related cerebral infarction have been described so far.[3–7] Three out of the five cases were from an apoplectic prolactinoma. This is the fourth documented case to date, according to a PubMed-indexed literature search.[3, 5, 7] Here, we report the first successful non-surgical management of acute ischemic stroke in an African adolescent male caused by an apoplectic prolactinoma

CASE REPORT

A 13-year-old male with a 2-year history of intermittent headaches and a progressive decrease in visual acuity presented to the pediatric emergency unit with an abrupt worsening of headaches. The headache was global, constricting, and graded 5/10 in severity. It was associated with repeated episodes of vomiting, palpitations, easy fatiguability, and galactorrhea. He had no chronic medical and illness. There was a family history of essential hypertension but no history of stroke or tumors.

He presented with a Glasgow Coma score (GCS) of 15/15 with no motor deficits. There was reduced acuity in the right eye, with left hemianopia and diplopia. There was no papilledema. The hormone profile showed elevated prolactin and low cortisol levels. (Table 1)

A brain MRI showed a large expansile homogenously enhancing sella lesion with suprasellar extension. (Figure 1) He was started on bromocriptine 0.5 mg daily. On day 10 of admission, he developed a sudden onset of right hemiplegia and slurred speech. His level of consciousness declined to a

Submitted: 29 October 2024
Accepted: 24 December 2024
Published: 10 January 2025

GCS of 8/15 (Eye 1, Verbal 2, Motor 5). He had ptosis of the left eye and anisocoria, with a dilated and unreactive left pupil; right facial nerve palsy; and difficulty swallowing.

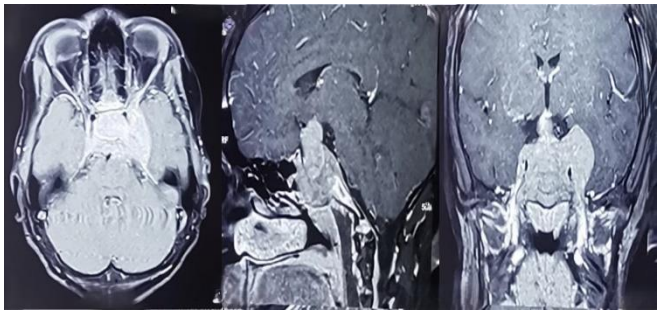

Figure 1- Brain MRI series upon admission shows a large, expansile, homogeneously enhancing sella lesion with supra-sella extension.

An impression of pituitary apoplexy was made. The patient was resuscitated and started on intravenous hydrocortisone. An urgent contrast-enhanced head CT showed a huge lobular sella mass with intra-tumoral hemorrhage and diffuse hypodensity in the left anterior cerebral artery and middle cerebral artery territories. (Figure 2)

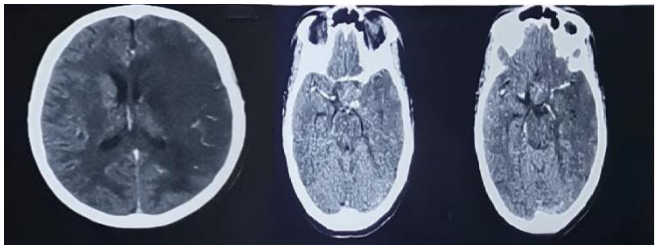

Figure 2 - Urgent Head CT done showed a large, (5.7ap x 6.2tr x 4.7) cm, non-enhancing, lobulated sella/suprasellar mass with hyperdensities of acute blood attenuation (average 54 HU) in the left aspect of the lesion, associated with effacement of the suprasellar cistern, compression of the optic chiasm, widening of the sella turcica and partial erosion of the clivus. Diffuse hypodensity is noted in the left anterior and middle cerebral artery (ACA, MCA) distributions.

Hormonal assays revealed markedly elevated prolactin, low cortisol, luteinizing, and thyroid hormone levels. (Table 1)

A diagnosis of pituitary apoplexy (PA) and an acute ischemic stroke in the left anterior and middle cerebral arteries was made. A trans-sphenoidal approach was considered as the initial decision for urgent surgical decompression. However, the lack of an Intensive care unit (ICU) space and ventilator for post-surgery recovery hindered the decision. After consulting with the endocrinologist, the team decided to optimize dopamine agonist therapy with cabergoline. Patient symptoms improved on medical management with cabergoline. There was also a significant decline in his serum prolactin levels when it was assessed 20 days after commencement of therapy.(Table 1)

Table 1. Patient Hormonal Level Trend: A- Done at diagnosis of pituitary apoplexy B- 20 day commencement of medical therapy

INVESTIGATION	A	B	REFERENCE
S- Prolactin	2973	36.68	2.64-13.13ug/L
S- Cortisol (AM)	30	24.70	166-507 nmol/L
S- ACTH*	1.5	N/A	2.8-64.6 pg/ml
S- TSH*	0.845	1.445	0.68-3.35 uIM/ml
S- T3*	4.9	1.92	3.98-6.19 pmol/L
IGF-1*	815.0		143-506ng/ml
S- LH*	4.18	0.59	1.24-8.62 IU/L
S- FSH*	2.36	1.66	1.27- 19.26 IU/L

Abbreviations: ACTH: adrenocorticotrophic hor… Stimulating hormone, IGF: insulin like growth factor , LH: Luteinizing hormone FSH: Thyroid Stimulation hormone, T3(triiodothyronine)

The GCS improved to 15/15, and there was resolution of the dysphagia and facial nerve palsy. There was no change in the other neurologic findings. A repeat MRI showed a reduction in tumor volume (Figure 3). On follow-up assessment one month after discharge, GCS was 15/15, and his right motor strength had improved with power in the right upper and lower limbs at 1/5.

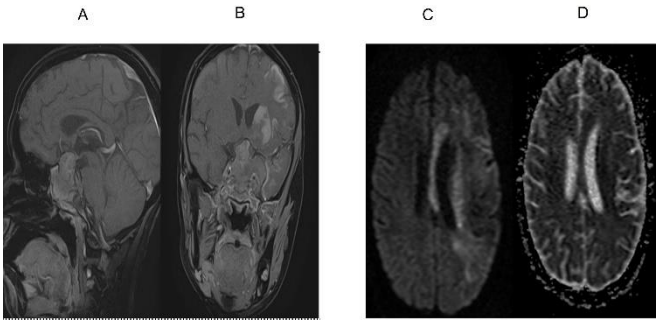

Figure 3- Brain MRI showing a reduction in size of the large, heterogeneously enhancing sella /supra sellar mass (A, B) to (4.2ap x 5.3tr x 4.2cc) cm, with persistent areas of subacute hemorrhage (T1 hyperintense, T2 hypointense), central focus of restricted diffusion and blooming artifacts on T2*. The mass partly eroded the clivus and extended to the left middle cranial fossa and both sphenoid sinuses. It compressed the optic chiasm, floor of the third ventricle, and invaded the left cavernous sinus, encasing the left internal carotid artery (Knosp grade 4 classification). There was cortical enhancement of the left frontoparietal lobes, left corpus callosum and the left basal ganglia with associated restricted diffusion on the DWI/ADC (C, D) in the left ACA, MCA territories.

DISCUSSION

The relationship between pituitary apoplexy and ischemic stroke is not well understood. Possible pathophysiology under consideration are: the mass effect from large tumors can lead to reduced blood flow due to pressure exerted on nearby vessels, as illustrated in this case; vasospasm from apoplexy-induced inflammation; and direct tumor infiltration into nearby vessels. [8–10] The vessels of a pituitary adenoma exhibit morphological features indicating insufficient maturation, inadequate fenestration, and frequent basal membrane rupture. The presence of these structural anomalies can increase the

DOI: 10.46900/apn.v7i1.288

likelihood of spontaneous bleeding, potentially leading to apoplexy.[11]

Jiang and colleagues reported that 58.7% of cases affected the internal carotid artery (ICA), 32.6% the anterior cerebral artery (ACA), and 25% the middle cerebral artery (MCA). In their study, mechanical compression was the most prevalent cause, whereas cerebral vasospasm accounted for around 40% of cases.[6] Four out of the five cases of cerebral ischemia from pituitary apoplexy in children were attributed to arterial compression, with the middle cerebral artery being the most affected. [3–7] MRI confirmed the blockage of the left internal carotid artery, leading to a stroke in the anterior cerebral artery (ACA) and middle cerebral artery (MCA) regions in this case.

It can be crucial in these cases to perform a CT or MR angiogram upon admission to assess the patency of the intracerebral vessels after PA [12]. In locations with limited resources, this luxury may not be available for most cases. In the absence of an angiography, it remains uncertain whether vasospasm or intracranial artery compression occurs in cases of cerebral infarction caused by a pituitary stroke.

Due to the infrequency of this condition, selecting between medicinal and surgical care for these patients can be difficult. While medical treatment is frequently effective for managing prolactinomas, the specific criteria for determining when surgery is necessary have not been precisely defined. [4] Hoffman et al. conducted a study on 27 patients with pediatric pituitary prolactinoma. They found that dopamine-agonist treatment is effective and safe for reducing tumor volume in pediatric prolactinoma.[13] Breil et al. reported an 80% reduction in 11 pediatric prolactinomas treated with cabergoline throughout a 37-month median follow up time. [1] Maiter et al demonstrated in their study of 455 patients, that cabergoline was first choice and also effective in patients with bromocriptine resistance.[14] Yousem et al observed a significant ($P<0.01$) association between bromocriptine use and intratumoral hemorrhage in patients with prolactinoma.[15] However due to the relatively cheaper cost of bromocriptine compared to cabergoline, bromocriptine is the usual first line in our setting since it is cheaper.

Urgent surgical intervention is usually required to relieve pressure on the optic chiasm if there is a risk to vision or medical therapy fails.[13] Decompression of the tumor via a trans-sphenoidal approach was considered the first option in this case, however, limited intensive care space for post operative care was a major limitation. It was soon noticed however that patient continued to improve clinically on medical management. Surgery would still be considered, if there was additional clinical decline, failure to normalize prolactin levels, or inadequate reduction in tumor size with medical therapy alone. [7]

This is the second reported case of ischemic stroke from prolactinoma that was treated with dopamine agonist only, similar to a 15-year-old reported by Taemin et al who didn't have apoplexy. [7]

Strengths and Limitations

A major limitation to this case report was the unavailability of angiography studies to establish whether the cause of the stroke was from direct compression or cerebral vasospasm. Additionally, planned surgical decompression at the diagnosis of apoplexy was delayed due to unavailablity of ICU. However, in the limited resource settings, the successful conservative management highlights a potential alternative to surgical intervention in select cases.

Patient's Perspective

The evolution of the stroke symptoms was overwhelming for the parents of the patient and they feared the worst outcome. The team kept parents abreast with the working diagnosis and management path. The final decision not to have surgery was very accommodating for the parents especially when there was a steady recovery of the child with medical therapy.

CONCLUSION

Pituitary apoplexy-induced cerebral infarction is a rare occurrence, particularly in children, with only a few published cases. Determining the most appropriate treatment modality for such unique might be challenging. Our findings further illustrate the significance of clinicians having a heightened level of suspicion and an individualized multidisciplinary approach to treatment. Additional reports and research are urgently needed to investigate the condition and provide clear guidelines that will assist in the identification and treatment of future possible instances, even in settings with limited resources.

ACKNOWLEDGMENTS

The authors would like to thank all the staff of the Department of Neurosurgery, Korle Bu Teaching hospital and the Pediatric endocrinology team for their contributions in the successful management of the patient

DISCLOSURES

Ethical approval

This study was performed in line with the principles of the Declaration of Helsinki. Approval was granted by the local Ethics Committee, number:KBTH-IRB/000198/2024

Consent to participate

The patients gave consent to use their information and images for research purposes. *Consent for publication*

The patient gave consent to use his information and images for publication.

Conflict of interest

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Funding

This research received no specific grant from any funding agency in the public, commercial or not-for-profit sectors

CONTRIBUTIONS

-**Omane Acheamfour Okrah**: Conceptualization, Methodology, Project administration, Resources, Validation, Writing – original draft, Writing – review & editing
-**Emmanuel Kwadwo Adjei Osei**: Conceptualization, Investigation, Methodology, Project administration, Resources, Validation, Writing – original draft, Writing – review & editing
-**Michael Yankey**: Data curation, Methodology, Writing – original draft, Writing – review & editing
-Joseph Kebo Awindago: Conceptualization, Data curation, Validation, Writing – review & editing
-Emmanuel Jackson: Software, Validation, Visualization, Writing – review & editing
-Patrick Bankah: Resources, Supervision, Validation, Writing – review & editingSoftware, Validation, Visualization, Writing – review & editing

REFERENCES

1. Briet C, Salenave S, Bonneville JF, Laws ER, Chanson P (2015) Pituitary apoplexy. Endocr Rev 36:622–645. https://doi.org/10.1210/er.2015-1042
2. Brougham M, Heusner AP, Adams RD (1950) Acute Degenerative Changes in Adenomas of the Pituitary Body—with Special Reference to Pituitary Apoplexy. J Neurosurg 7:421–439. https://doi.org/10.3171/jns.1950.7.5.0421
3. Agrawal P, Newbold S, Al Busaidi A, et al. (2022) Pituitary apoplexy in an adolescent male with macroprolactinoma presenting as middle cerebral artery territory infarction. Ann Pediatr Endocrinol Metab 27:320–324. https://doi.org/10.6065/apem.2142204.102
4. Pozzati E, Frank G, Nasi MT, Giuliani G (1987) Pituitary Apoplexy, Bilateral Carotid Vasospasm, and Cerebral Infarction in a 15-Year-Old Boy. Neurosurgery 20:56–59. https://doi.org/10.1227/00006123-198701000-00016
5. Kasl RA, Hughes J, Burrows AM, Meyer FB (2015) Pediatric ischemic stroke from an apoplectic prolactinoma. Child's Nervous System 31:1387–1392. https://doi.org/10.1007/s00381-015-2712-8
6. Jiang Q, Xiao S, Shu L, Huang X, Chen X, Hong H (2020) Pituitary Apoplexy Leading to Cerebral Infarction: A Systematic Review. Eur Neurol 83:121–130. https://doi.org/10.1159/000507190
7. Oh T, Amara D, Gupta N, Clerkin P (2020) A middle cerebral artery ischemic stroke occurring in a child with a large prolactinoma. Child's Nervous System 36:853–856. https://doi.org/10.1007/s00381-019-04446-z
8. Wortham J, Sandoval B, Koops M, Granda-Rodriguez R, Bruder JM (2021) Giant Prolactinoma Causing Proptosis and Stroke. J Endocr Soc 5:A584–A584. https://doi.org/10.1210/jendso/bvab048.1191
9. Iglesias P (2024) Pituitary Apoplexy: An Updated Review. J Clin Med 13:2508. https://doi.org/10.3390/jcm13092508
10. Gambaracci G, Rondoni V, Guercini G, Floridi P (2016) Pituitary apoplexy complicated by vasospasm and bilateral cerebral infarction. BMJ Case Rep bcr2016216186. https://doi.org/10.1136/bcr-2016-216186
11. Benigno A, Santos A, França MM, et al. (2011) Conservative management of pituitary tumor apoplexy Tratamento conservador da apoplexia pituitária clinical case report. https://doi.org/10.1590/S0004-27302011000500008
12. Navarro-Bonnet J, Martínez-Anda JJ, Balderrama-Soto A, Pérez-Reyes SP, Pérez-Neri I, Portocarrero-Ortiz L (2014) Stroke associated with pituitary apoplexy in a giant prolactinoma: a case report. Clin Neurol Neurosurg 116:101–103. https://doi.org/10.1016/j.clineuro.2013.09.039
13. Hoffmann A, Adelmann S, Lohle K, Claviez A, Müller HL (2018) Pediatric prolactinoma: initial presentation, treatment, and long-term prognosis. Eur J Pediatr 177:125–132. doi: 10.1007/s00431-017-3042-5
14. Maiter D (2019) Management of Dopamine Agonist-Resistant Prolactinoma. Neuroendocrinology 109:42–50. https://doi.org/10.1159/000495775
15. Yousem DM, Arrington JA, Zinreich JS, Kumar AJ, Bryan NR (1989) Pituitary adenomas possible role of bromocriptine in intratumoral hemorrhage. Radiology 170:239–243. https://doi.org/10.1148/radiology.170.1.290